ABSTRACT

Lichen planus pigmentosus is a photodistributed dyschromia of unknown etiology described clinically as hyperpigmented gray-blue or brown-black macules or patches in a photo-distributed pattern. Although there has been some debate, lichen planus pigmentosus is considered by many to be a separate diagnostic entity from ashy dermatosis or erythema dyschromicum perstans, which shares similar characteristics. Various treatment strategies have been applied to help resolve or improve the appearance of lichen planus pigmentosus lesions; however, an optimal treatment method is yet to be elucidated. The authors present a case of an 18-year-old Hispanic man with lichen planus pigmentosus whose skin findings responded dramatically to a combined regimen of daily topical azelaic acid foam and tretinoin cream with twice-monthly chemical peels using glycolic acid and Jessner's solution. The authors have noted a sparcity of therapeutic literature for lichen planus pigmentosus, and hope to aid clinicians in therapeutic management strategy for this patient subset.

*J Clin Aesthet Dermatol.* 2016;9(11):44–50

CASE REPORT

# A Case of Lichen Planus Pigmentosus with Facial Dyspigmentation Responsive to Combination Therapy with Chemical Peels and Topical Retinoids

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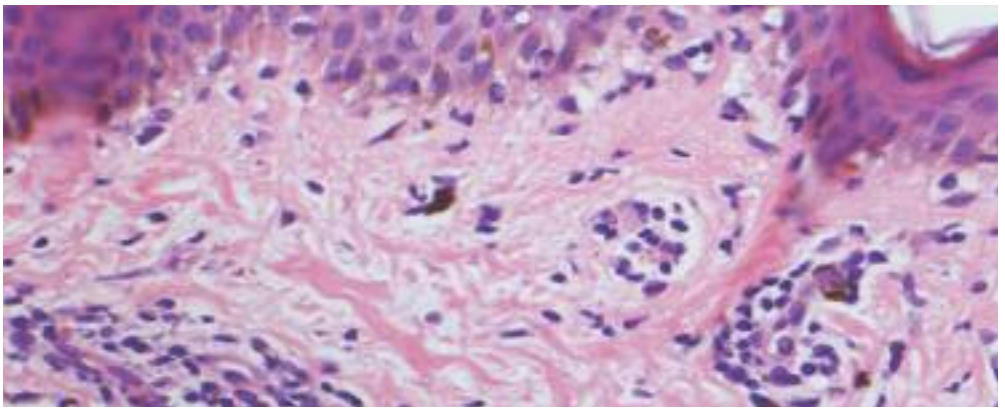

LICHEN PLANUS PIGMENTOSUS (LPP) is considered a macular variant of lichen planus with an unknown etiology. The condition was first described by Bhutani et al as a condition recognized in India, presenting in the third to fourth decade of life and manifesting as dyspigmented macules or patches.[1–4] Since its discovery, LPP has often been reported in a variety of ages and darker skin typed ethnicities including Hispanics and African Americans.[4,5] Lesions can exhibit a range of pigmentary dyschromia, from brown-blackish to blue or purple-gray macules and patches, which predominantly involve the sun-exposed aspects of upper extremities, face, and neck.[4] Areas of involvement tend to be asymptomatic, but pruritus or burning may be present.

Histology classically demonstrates hyperkeratosis, epidermal thinning,

**Disclosure:** The authors report no relevant conflicts of interest.

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vacuolar degeneration of the basal cell layer, lymphohistiocytic band-like infiltrate, variable Civatte or colloid bodies, and melanin incontinence.[3–5] There are several treatment options that have been reported with variable efficacy for improvement of the appearance of the lesions, but due to the chronicity of the underlying disease process, none of these treatments are curative.

## CASE REPORT

An 18-year-old Hispanic man presented to our Bronx, New York, clinic in autumn with an eight-month history of an asymptomatic dyspigmentation of the skin on his face, neck, and bilateral forearms that had gradually worsened. He denied preceding cutaneous redness, scale, pain, or pruritus. There was no truncal or lower extremity involvement, but he did admit to frequent sun exposure and lack of sun protection. Past medical history was significant only for mild facial acne, with no history of topical prescription or over-the-counter medications. No one in his family had similar lesions, and he denies any personal or family history of autoimmune disease.

Physical examination demonstrated multiple round-to-oval blue-gray macules and patches with some areas of coalescence scattered symmetrically on lateral aspect of his cheeks, neck, and circumferentially on his forearms in a photodistributed pattern (Figures 1 and 2).

Differential diagnosis included LPP, erythema dyschromia perstans, drug-induced hyperpigmentation (although denied minocycline use), post-inflammatory pigmentary alteration from a resolved dermatitis, pigmented contact dermatitis, and a resolved phototoxic or photoallergic reaction.

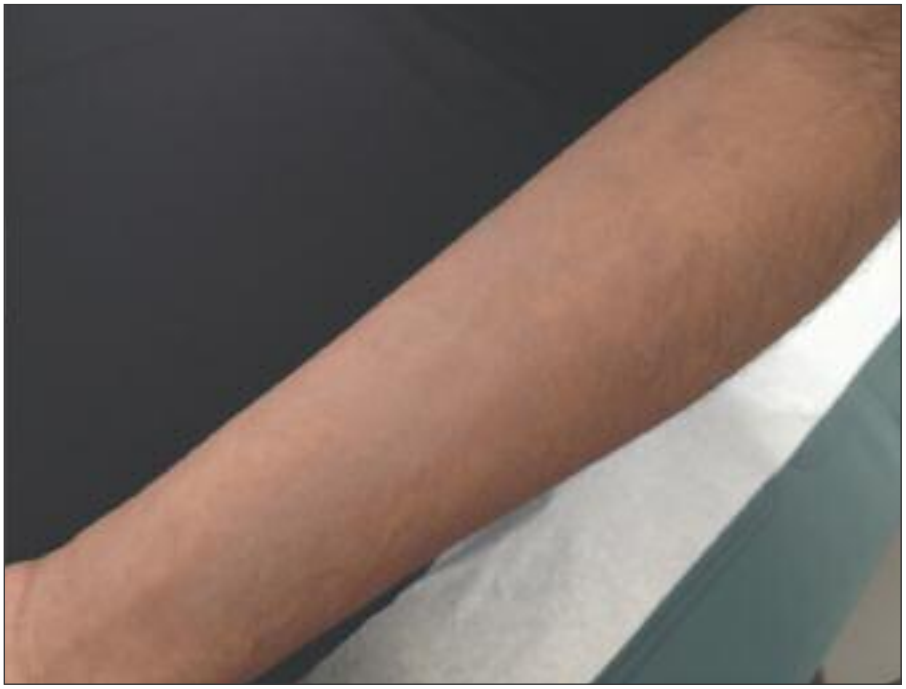

**Figure 1.** Multiple round-oval blue-gray macules coalescing into patches on right forearm.

A 3mm punch biopsy obtained from the right forearm demonstrated a patchy vacuolar interface dermatitis with melanin incontinence, dermal melanophages, and mild dermal atrophy (Figure 3). These findings, in the setting of his clinical presentation, confirmed the diagnosis of lichen planus pigmentosus.

Initial treatment regimen included daily sun protection with use of sunscreen with a minimal sun protection factor (SPF) of 30 and protective clothing. Following biopsy confirmation of the diagnosis, combination therapy was added with topical lightening medications and superficial chemical peels for lightening and resurfacing. The authors began the patient on topical 5% azelaic acid foam to face to be applied in the morning, and tretinoin 0.1% cream to face and arms at night. Chemical peels were applied every two weeks, with Jessner's peel selected for his arms and glycolic acid peels applied to his face. Jessner's peel requires no neutralization and was evenly applied to entire upper extremities with one layer at initial visit, with two layers at each follow-up. Glycolic acid peels were administered evenly with two passes on the entire face, sparing only periocular skin and ears.

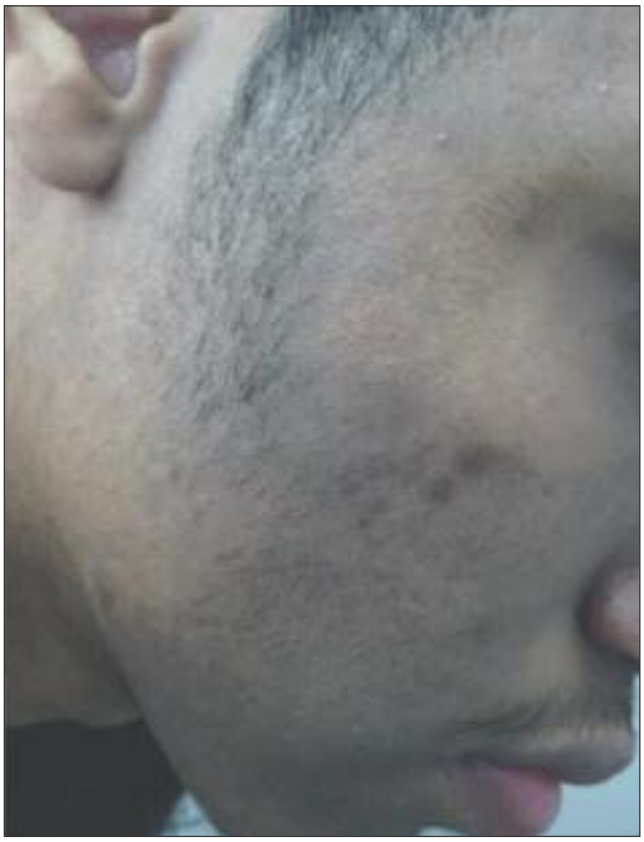

**Figure 2.** Multiple round-oval blue-gray macules coalescing into patches along the temple, cheek, jaw, and neck.

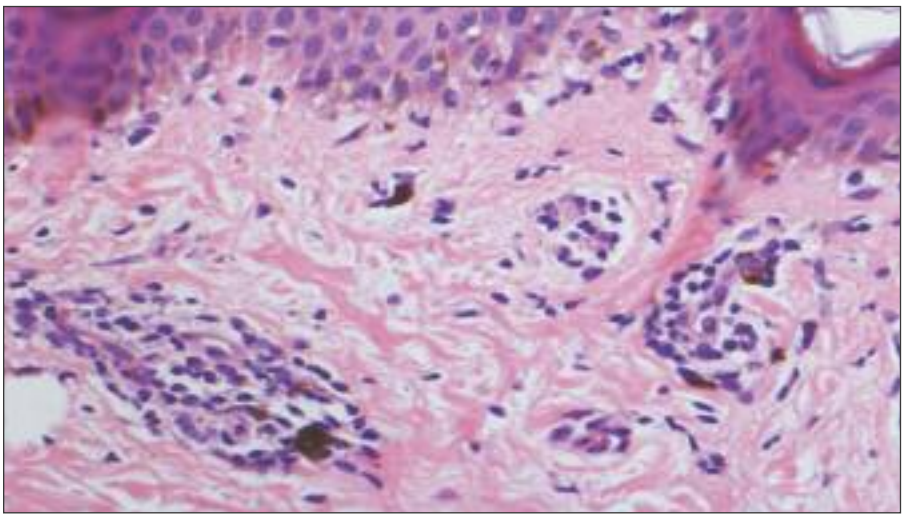

**Figure 3.** H&E (40x) revealing patchy vacuolar interface dermatitis with melanin incontinence, dermal melanophages, and mild dermal atrophy.

Following an even application, the patient was under physician observation for redness, itching, burning, or discomfort to signify endpoint of treatment of glycolic acid, with time ranging from 2 to 8 minutes as tolerated symptomatically by the patient. At endpoint, the glycolic acid was neutralized with water, and sunscreen was applied to face and arms. Glycolic peel strength was increased sequentially at each visit, with two treatments of 35% followed by two treatments of 50% glycolic acid. Peels had been premade by the manufacturer and were not buffered or altered. After eight weeks of therapy (four peels), the authors' patient noticed a significant improvement in his facial dyspigmentation (Figure 4). Peel frequency then changed to monthly due to schedule limitations, and topical therapy and photoprotection measures were continued daily. He received two additional chemical peels with Jessner's for his arms, and glycolic acid 70% on his face. After 16 total weeks of treatment, a marked improvement in the appearance of his facial lesions was noted (Figure 5). While the dyschromia on his arm lesions did mildly improve, the difference was not as clinically dramatic as his facial response. Future therapy will include neodymium-doped yttrium aluminium garnet (Nd-YAG) laser and the addition of tacrolimus ointment.

## DISCUSSION

Lichen planus pigmentosus is a chronic relapsing and remitting

dyspigmentation of the skin that has primarily been identified in darker ethnic skin types commonly in young-to-middle-aged adults, particularly in the third to fourth decade.[3,5] LPP was first described in 1974 by Bhutani et al after they investigated a subset of Indian patients with similar dermatologic findings to erythema dyschromicum perstans (EDP) (also known as ashy dermatosis), except that they also had associated lichen planus clinically and histologically.[5] They considered this new entity as a macular variant of lichen planus. While there is considerable overlap between EDP and LPP, in 1992, Vega et al[6] reported that these conditions are distinct entities and presented clinical and histopathological differences between the two. Similarities in the physical and histologic appearance of LPP and EDP have made the diagnosis difficult in the past, but notable differences identified by Vega et al has helped clinicians in distinguishing between the two diseases.[6]

Both LPP and EDP are characterized by dyschromic dark-brown or slate-gray macules. Distinction can be made early on, as lesions of EDP initially present with an asymptomatic dyschromia with an erythematous and slightly elevated margin and often involve sun-protected areas such as the trunk and proximal extremities, but can also include the face, neck, and upper extremities.[6–8] Within several months, this erythematous margin seen in EDP will disappear. Alternatively, the lesions of LPP

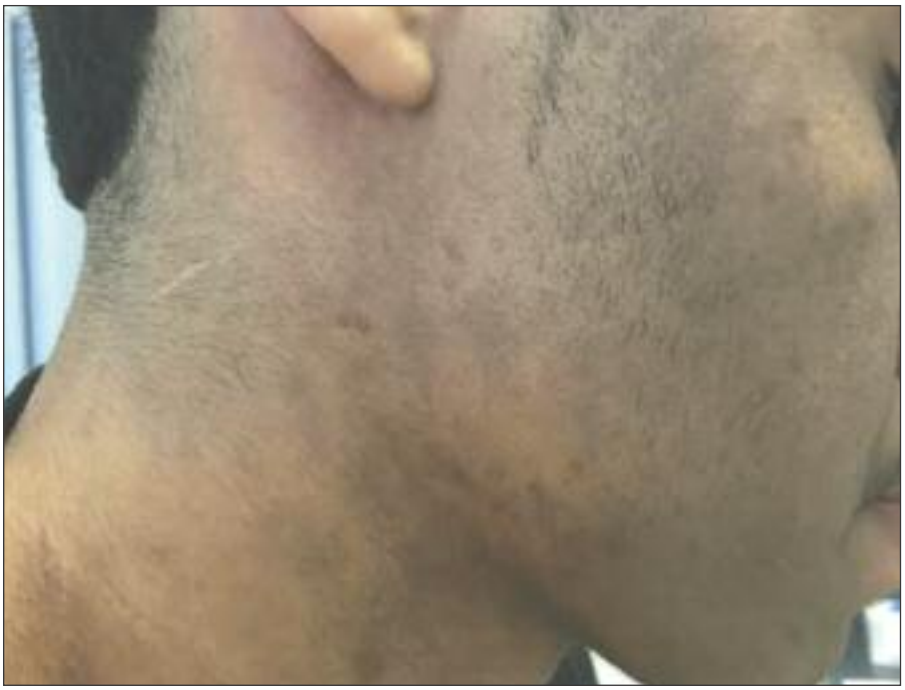

**Figure 4.** Decreased blue-gray dyschromia of face after four weeks of biweekly superficial glycolic acid chemical peels and daily azelaic acid foam and tretinoin cream.

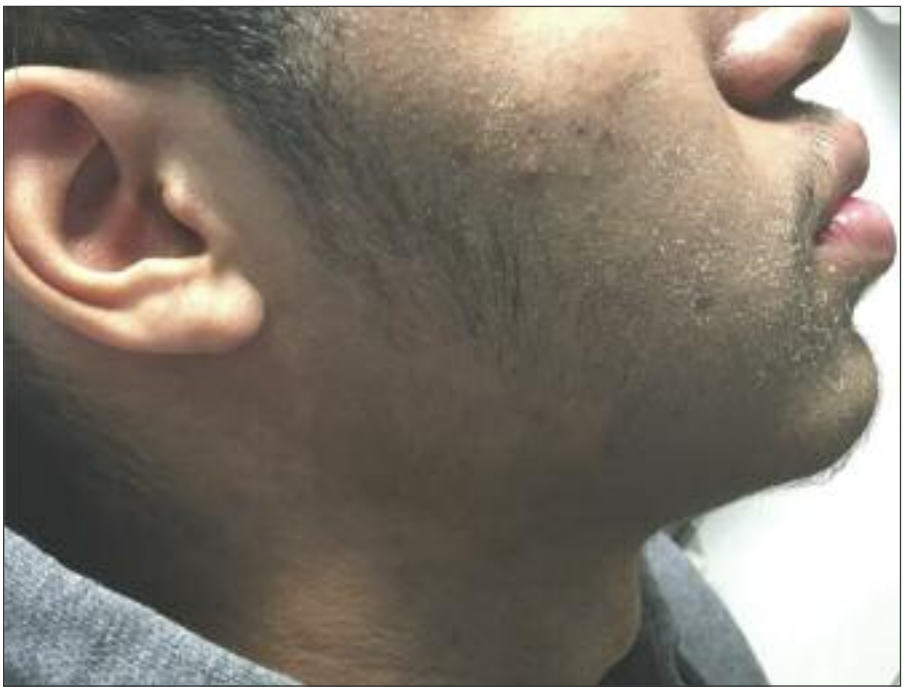

**Figure 5.** Markedly improved dyschromia of face and neck after 16 total weeks of therapy.

lack erythematous changes that are seen in EDP, can be pruritic (unlike EDP), and usually effect sun-exposed areas of the face, neck, and proximal flexural extremities.[6–9] It has been reported that approximately 20% of patients with LPP also have classic lichen planus disease.[6,10] Both diseases are associated with a chronic and insidious course. Histopathologic overlap is seen between the two conditions with shared features of epidermal atrophy, vacuolar interface dermatitis, and pigment incontinence with dermal melanophages.[6] When present in biopsy specimen, a lichenoid infiltrate, hyperkeratosis, and hypergranulosis can aid in the diagnosis of LPP. Lichenoid interface reactions and colloid bodies are more pronounced in LPP and only occasionally seen in EDP.[11] Interface lymphohistocytic infiltrate is more prominent in EDP, particularly in the early erythematous phase, however these changes can be present in both entities.[6]

Lesions of LPP initially appear as small, ill-defined, oval-to-round macules, which later become confluent to form large areas of pigmentation. Pigmentation varies from slate gray to blue to brown to brownish-black; in a single patient, the pigment is generally uniform. The face and neck are the most frequent initial sites, followed by the upper extremities.[2] Coexistent classic lichen planus lesions can be seen in up to 20 percent of LPP patients.[11]

Photodistributed dyschromias often occur in darker skin types (Fitzpatrick types IV–VI). This hyperpigmentation or dyspigmentation in areas of normal sun exposure are classically seen in conditions such as lichen planus pigmentosus, erythema dyschromia perstans, photoaggravated atopic dermatitis, drug-induced photosensitivity, pigmented contact dermatitis, and actinic lichen planus.[1,2] Widespread or localized hyperpigmentation is associated with varieties of conditions and may be due to genetic or systemic factors, such as metabolic, endocrine, chemical (drug), physical, nutritional, postinflammatory, or neoplastic disease.[11] While clinical history and histologic findings can help narrow the differential diagnosis, similar clinical and histologic findings between LPP and pigmented contact dermatitis have been reported. Distinguishing features of pigmented contact dermatitis is that it often involves the forehead and is preceded by erythema, papules, swelling, and itching, which lead to gradual hyperpigmentation by frequent and repeated contact with very small amounts of the contact sensitizer.[11] Contact sensitizers commonly implicated are those within the cosmetic, textile, fragrance, and dye categories. Unlike LPP, pigmented contact dermatitis will resolve once contact with the allergen is discontinued, while LPP will wax and wane.

Pigmentary dyschromias occur in greater frequency and severity in darker skin types and are a frequent reason for dermatologic consultations. Inflammatory skin reactions all have the ability to disturb the dermoepidermal junction, causing melanin to pass into the dermis and producing persistent hyperpigmentation that can make treatment difficult.[10] Standard initial therapy for any dyschromia includes the removal of aggravating factors, vigorous photoprotection, and a form of active pigment reduction with topical agents or physical modalities.[11] Sun avoidance when possible, and diligent photoprotection with sunscreen and sunblock is of particular importance in dark-skinned individuals, as they may be unaware of the darkening effects of ultraviolet (UV) radiation and the exacerbation of hyperpigmentation.[11] Supplementation with daily 1000 IU vitamin D is an important consideration as this patient population is also at risk of developing a deficiency.[11]

The pathophysiology behind lichen planus-related skin disease classically involves a T-cell mediated, autoimmune inflammatory response. Although the exact etiology of LPP remains unknown, viral infections, topical agents including mustard oil, amla oil, and henna hair dye, as well as sunlight have been shown to incite reactions; avoidance of these triggers is recommended.[3] Topical agents include hydroquinone, which is the most commonly used agent for pigmentary dyschromia, often in combination with topical retinoic acid, corticosteroids, azelaic acid, kojic acid, and

glycolic acid. Additional reported modalities including lightening agents, such as arbutin, niacinamide, N-acetylglucosamine, ascorbic acid, licorice, and soy, require further, larger controlled studies examining their efficacy.[1]

Literature supporting therapeutic options for LPP have shown inconsistent efficacy and are limited to isolated case reports and few, small-investigative case series. Reported modalities for LPP therapy include topical glucocorticoids, topical immunomodulators, topical keratolytics, superficial chemical peels, oral steroids, oral vitamin A, and Q-switch Nd-YAG laser therapy. Oral clofazimine and dapsone have been reported in EDP cases, but have not been reported in LPP.

In a study treating LPP with daily 100,000 IU vitamin A for 15 days followed by 15 days without treatment, some participants noticed a good-to-excellent improvement in hyperpigmentation after 10 or more treatments.[4] Systemic vitamin A is believed to act as a lysosomal labilizer and can break melanin in the melanophages of the upper dermis into smaller particles.[4]

An open label, nonrandomized study by Al-Mutairi et al [12] reported about half (13 out of 30) of the patients treated with twice-daily application of 0.03% tacrolimus ointment showed some lightening of the pigmentation over an average of 12 weeks.The mechanism behind topical calcineurin inhibitors as an effective therapy can be attributed to modulation of an abnormal cell-mediated immune response, which has been postulated to play a role in the pathogenesis of both LPP and EDP.[9,10] Topical calcineurin inhibitors (tacrolimus, pimecrolimus) bind with cytoplasmic immunophilin FKBP12 and form a complex that inhibits the activity of the enzyme calcineurin needed for T-cell activation. While tacrolimus ointment may be helpful in treating LPP, due to limited data and inconsistent efficacy, more definitive treatment options are required.

In 2014, Han et al[13] reported a case study of a patient responding to low-fluence Q-switch Nd:YAG laser therapy. This patient initially failed topical therapy with topical betahydroxy acids and hydroquinone-tretinoin-fluocinolone formulations. This patient received low-fluence Q-switched Nd:YAG laser every two weeks for 28 sessions.[13] A 6mm spot using a fluence of 3J/cm$^2$ and pulse duration of 5ns was used on the cheek for 10 passes.[13] One year following treatment, her cheek maintained the improvement from the laser with very minimal macular pigmentation.[13] Laser therapy may serve as a promising therapeutic option in this subset of patients, and further investigation with larger case series or controlled studies would be beneficial as evidence-based data are currently lacking.

Chemical resurfacing with superficial glycolic acid and Jessner's peel was chosen as the initial treatment for the authors' patient in combination with daily lightening and exfoliative topical agents of azelaic acid and tretinion. The goal of these modalities was to reduce further dyschromia and to induce controlled injury to the epidermis up to the dermal-epidermal junction with hope to encourage keratinocyte turnover and promote melanophage clearance. Strict photoprotection with daily facial sunscreen with a minimal SPF of 30 and long-sleeved shirts were followed by the patient, with rationale being to prevent further darkening of his dyspigmentation by decreasing UV-induced melanogensis. Adherence with daily topical 5% azelaic acid foam and nightly tretinoin 0.1% cream were endorsed over the 16-week treatment period, and the patient had significant improvement of facial lesions. Future therapeutic options the authors plan on adding to the patient's therapy are tacrolimus cream and Nd:YAG laser. While the patient has noticed a decrease in pigmentation with his current regimen, the authors hope the addition of these modalities will also play a role in improving the appearance of the lesions.

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